[DOI]10.12016/j.issn.2096-1456.2020.02.006

·防治实践·

# 下颌下腺导管畸形伴多发结石病例报道及文献复习

李瑞春[1]，景凯岸[2]，王丽娜[1]，马瑞[2]，景捷[2]，马坚[2]
1. 宁夏医科大学口腔医学院，宁夏 银川（750000）；2. 宁夏医科大学总医院口腔颌面外科，宁夏 银川（750000）

**【摘要】 目的** 探讨下颌下腺导管畸形伴多发结石的诊治方法。**方法** 对1例罕见的下颌下腺导管畸形伴多发结石患者的临床资料、诊断及治疗进行总结，并回顾相关文献。**结果** 该患者查体及CBCT检查发现左侧口底区肿物，通过术中探查发现患者左侧口底肿物为下颌下腺导管畸形伴多发结石形成，切除下颌下腺及肿物。术后病理检查报告为：（左侧下颌下）涎腺组织导管扩张并导管上皮增生，可见导管结石，导管周围大量淋巴细胞增生，术后1月随访愈合良好。通过文献回顾分析发现，下颌下腺导管畸形伴多发结石的病例较少见，应与口底区动静脉畸形仔细鉴别。下颌下腺导管结石可以经口行下颌下腺导管切开取石或经颌下行下颌下腺并导管结石切除，对一些直径较小的结石也可通过唾液腺内镜治疗。**结论** 下颌下腺导管畸形伴多发结石的病例需同时行口内及口外切口切除下颌下腺并导管结石。

**【关键词】** 下颌下腺；下颌下腺导管；导管结石；导管畸形；多发结石；鉴别诊断；口底区动静脉畸形；治疗；预后
**【中图分类号】** R781.7 **【文献标志码】** A **【文章编号】** 2096-1456（2020）02-0093-04
开放科学（资源服务）标识码（OSID）
**【引用著录格式】** 李瑞春，景凯岸，王丽娜，等. 下颌下腺导管畸形伴多发结石病例报道及文献复习[J]. 口腔疾病防治，2020，28(2)：93-96.

**Malformation of Wharton′s duct with multiple sialoliths: a case report and literature review**

LI Ruichun[1], JING Kaian[2], WANG Lina[1], MA Rui[2], JING Jie[2], MA Jian[2]. 1. Oral Medicine School of Stomatology, NingXia Medical University, Yinchuan 750000, China; 2. Department of Oral & Maxillofacial Surgery, General Hospital of Ningxia Medical University, Yinchuan 750000, China.
Corresponding author: MA Jian, Email: majianhs310@163.com, Tel: 86-951-6743384

**【Abstract】 Objective** To explore the diagnosis and treatment of ductal malformations of the submandibular gland with multiple stones. **Methods** A case of a malformation of Wharton′s duct with multiple sialoliths according to the clinical data, diagnosis and treatment of the patient was analyzed retrospectively. **Results** The patient′s physical examination and CBCT showed a tumor on the left floor of the mouth. In this case, it was found that the mass was a malformation of Wharton′s duct with multiple sialoliths according to operative exploration. The postoperative pathological examination showed (left submaxillary) salivation gland tissue, duct dilation and duct epithelia hyperplasia, duct calculus, and a large number of lymphocytes proliferating around the duct; 1 month after the follow-up, the patient had healed well. Through literature review and analysis, it was found that cases of submandibular ductal malformation with multiple stones were rare and should be carefully differentiated from arteriovenous malformation at the base of the mouth. Calculi of the submandibular gland can be removed by incision through the oral submandibular duct or by combined resection of the submandibular gland and ductal calculi, and some smaller calculi can also be treated by endoscopy of the salivary gland. **Conclusion** In cases of submandibular ductal malformation with multiple stones, intraoral and extraoral in-

**【收稿日期】** 2019-02-11；**【修回日期】** 2019-07-22
**【基金项目】** 国家自然科学基金项目（81600853）；宁夏高等教育科学研究项目（NGY2016121）
**【作者简介】** 李瑞春，住院医师，硕士研究生在读，Email：719952158@qq.com
**【通信作者】** 马坚，副主任医师，博士，Email：majianhs310@163.com, Tel: 86-951-6743384

cisions should be performed simultaneously to remove the associated ductal stones.
【Key words】 submandibular gland; Wharton's duct; ductal stones; duct abnormalities; multiple sialoliths; differential diagnosis; arteriovenous malformation in the oral area; treatment; prognosis
**J Prev Treat Stomatol Dis, 2020, 28(2): 93-96.**

唾液腺结石是腺体或导管内发生钙化团块引起的一系列病变，约87%发生于下颌下腺，且多发于下颌下腺导管[1-2]。多数下颌下腺结石病例为单发结石，多发结石者约占15%～20%[3]。下颌下腺结石好发年龄在30～60岁[4]。结石位于腺体外导管者占60%～70%，位于腺门或腺内导管者占30%～40%[5]。已有研究报道的下颌下腺导管结石最多有11枚[6]，本病例报道1例罕见的下颌下腺导管结石患者，其结石数目多达32枚，结石位于下颌下腺导管畸形处，现报道如下。

## 1　病例资料

### 1.1　一般资料

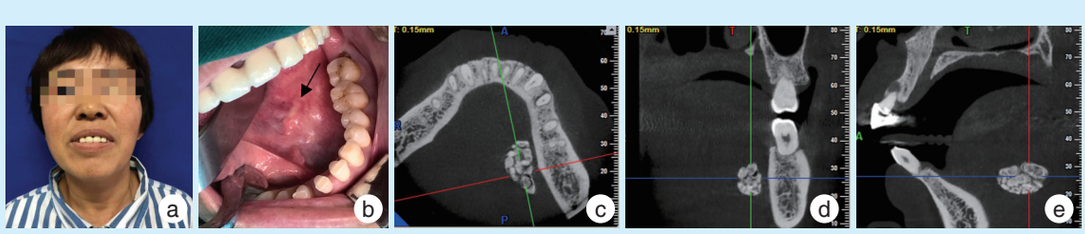
患者，女，49岁，因"无意中发现左侧口底区肿物2周"就诊。自诉2周前于当地医院拍牙科X线片时无意中发现左侧口底区肿物，无不适。否认创伤及左侧下颌下区口底肿胀病史。现为进一步治疗来宁夏医科大学总医院就诊。

### 1.2　临床检查

患者双侧面部轮廓基本对称，双侧下颌下区未见肿胀（图1a）；口内：开口度，开口型正常，36～37口底区黏膜较对侧稍隆起（图1b箭头所示），表面黏膜颜色及温度正常，可触及一大小约1.5 cm×1.0 cm肿物，压痛（-），质硬，界清，其内可触及多个大小不等、可活动的硬结性肿块，并可闻及肿块间摩擦音，似"沙包"样感觉，双合诊检查双侧下颌下腺质软，导管口分泌正常；双侧下颌下区及颈部未触及明显肿大淋巴结。

### 1.3　辅助检查及诊断

锥形束CT（cone beam CT，CBCT）检查发现35～37口底区多个大小不等高密度团块影，大小约1.5 cm×1.0 cm（图1c～e）。B超检查示：双侧下颌下腺大小形态正常，包膜完整，腺体回声均匀，未见占位病变。血常规、生化、凝血和胸片、心电图等检查结果均正常。术前诊断：左侧口底肿物。根据患者的查体和影像学检查考虑该肿物为导管结石可能性大。




a：frontal view；b：intraoral view；c：orizontal position；d：coronal position；e：sagittal position

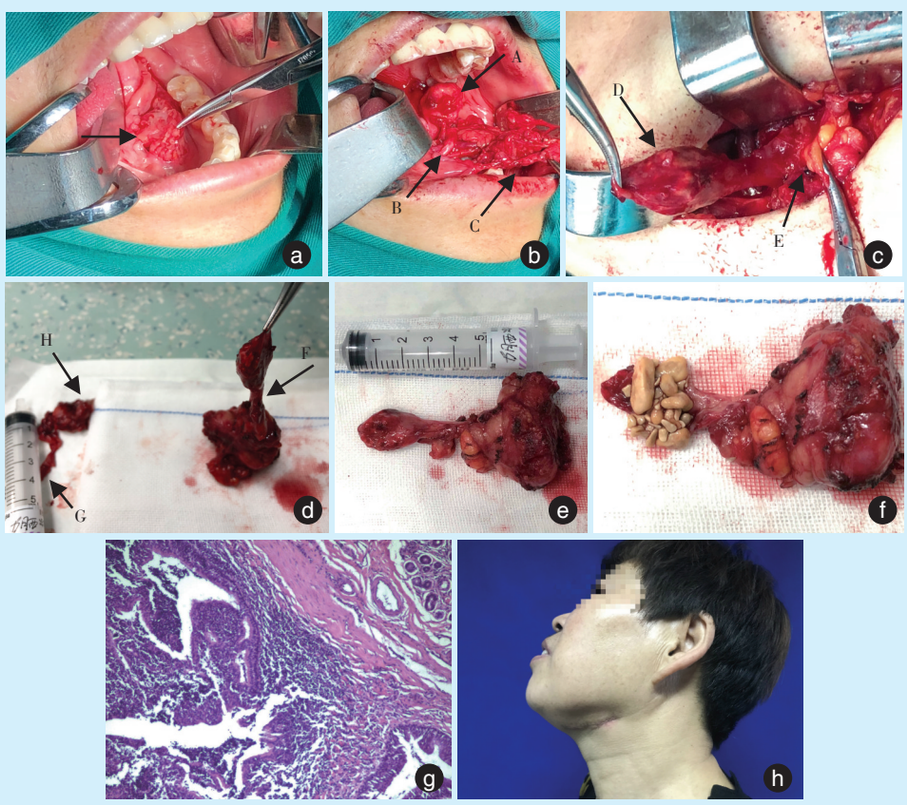
Figure 1　Preoperative views and CBCT images of the patient

**图1**　患者术前照片及CBCT图像

## 2　结　果

本院对患者全麻下行"左侧口底肿物探查术"。术中自肿物黏膜表面做一长约5.0 cm切口，切开黏膜及黏膜下层，分离提起舌下腺内侧缘（图2a），见肿物位于舌下腺深面；分离周围组织将肿物完整暴露，提起肿物，见肿物有蒂样结构位于舌神经深面；继续向深面分离，见其与下颌下腺相连，故该蒂样结构为下颌下腺导管，导管基部见分支导管样结构2支（直径约2 mm）与舌下腺导管汇合开口于舌下肉阜（图2b～d）；因该肿物较大，且导管基部较短，无法将下颌下腺导管异位开口，与患者家属沟通并征得同意后口外切口切除下颌下腺并肿物（图2c～e），因术中为了充分暴露肿物并寻找下颌下腺导管而损伤舌下腺导管及腺体，为避免后期形成舌下腺囊肿，与患者家属沟通并征得同意后一并摘除左侧舌下腺（图2d～f）。

术后病理检查提示:(左侧下颌下)涎腺组织导管扩张并导管上皮增生,可见导管结石,导管周围大量淋巴细胞增生(图2g)。术后1月随访,患者无明显不适主诉,口内外切口愈合良好(图2h)。




a: the arrow in the picture refers to the location of the tumor; b: arrow A in the picture refers to the tumor, arrow B refers to lingual nerve, arrow C refers to branch conduit; c: arrow D refers to the tumor, arrow E refers to submandibular gland; d: arrow F refers to the basal branch of submandibular duct, arrow G refers to the branching duct of submandibular gland and the part connected with sublingual gland, arrow H refers to sublingual gland; e: excision of submandibular gland with tumor; f: we can see about 0.5-1.0 cm in diameter and 32 sialoiths with smooth surface in the open tumor and duct; g: postoperative pathological examination (HE × 100) it can be seen that the duct were dilated and epithelia proliferated, and a large number of lymphocytes infiltrated around the duct; h: postoperative recovery

Figure 2　Surgical procedure and postoperative situation

**图2**　手术过程及术后情况

## 3　讨　论

一般认为下颌下腺之所以好发结石,是因为下颌下腺位于导管开口以下,涎液逆重力方向排泄;导管系统粗大、弯曲,异物易于进入导管;下颌下腺分泌的涎液粘稠,钙磷等矿物质浓度高,易沉淀发生结石[2]。多数的下颌下腺结石病例临床表现有明确的阻塞症状,并与进食关系密切[7]。

本病例中患者无左侧口底肿胀史,并且肿物位于口底肌以上,查体可触及,位置较表浅,术前需与口底血管畸形产生的静脉石相鉴别[8]。根据体位实验[9]、肿物表面覆盖黏膜的颜色[10]、核磁增强检查和下颌下腺导管走形方向可以与之有效鉴别。本病例中肿物所在表面黏膜颜色正常,未呈蓝色,体位实验阴性,并且高密度团块影位于下颌下腺导管走行区域,所以考虑结石可能性大。术中探查肿物位于舌下腺深面,完整暴露肿物发现其为一盲袋结构,包膜完整,其有蒂位于舌神经深面与下颌下腺相连,考虑该蒂样结构为下颌下腺主导管,导管基部见分支导管样结构2支(直径约2 mm)与正常下颌下腺导管走形相一致并与舌下腺导管汇合开口于口底,使得下颌下腺分泌的大量唾液经分支导管排入口内,所以患者左侧下颌

下腺无肿胀症状；由于大量唾液经主导管流至盲袋内，唾液流速减慢，积聚于该盲袋内，造成大量钙盐沉积，易形成多发结石，所以本病例结石数目多达32枚；同时由于导管通畅，大量唾液具有冲刷作用，使得结石表面光滑且大小不等。

口腔颌面部畸形多见于动静脉畸形，发生于唾液腺导管的畸形多以腮腺导管为主，发生于下颌下腺导管的畸形非常少见，国内外文献对此类病例鲜有报道。Prosdocimo等[11]曾报道1例先天性下颌下腺导管囊性扩张的病例，表现为口底区囊肿样变。一般认为下颌下腺导管囊性扩张是因为结石造成下颌下腺唾液分泌受阻[12]，但薛传鹏等[13]曾报道了4例下颌下腺导管囊性扩张误诊为舌下腺囊肿的病例，并不是由结石造成，作者推测也有可能是导管先天解剖变异引起。因此，由于下颌下腺导管畸形临床少见，术前需要对患者病史进行详细了解，根据患者的病史、查体、辅助检查，对患者病情以及手术方案进行充分的预估。通常下颌下腺结石的治疗方法有：口内下颌下腺导管切开取石，下颌下腺结石并腺体切除，也有利用唾液腺内镜下经口摘除下颌下腺结石的报道，只是该方法适用于直径较小的结石[14-15]。体外冲击波碎石术和唾液腺内镜下导管内激光碎石术，适用于唾液腺内镜下无法取出的大结石，这些方法取得了一定效果，但尚待积累更多经验[12]。

本文报道的病例临床上少见，是下颌下腺导管畸形所致的多发结石。病例总结：①由于畸形的下颌下腺主导管管径粗大，分支导管位于导管基部且分支导管管径细小，若结扎主导管保留下颌下腺，术后易引起结扎处盲端膨大及涎液潴留更易形成结石并腺体炎症可能，所以该类病例不建议保留下颌下腺；②本病例术中探查时为了充分暴露肿物并寻找下颌下腺导管而损伤舌下腺导管及腺体，为避免后期形成舌下腺囊肿，与患者家属沟通并征得同意后一并摘除左侧舌下腺，所以该类病例口内切口尽量避免损伤舌下腺，口内切口以探查下颌下腺畸形导管及结石为主，行口外切口切除下颌下腺及畸形导管和结石。

## 参考文献

[1] Kopeć T, Wierzbicka M, Kałużny J, et al. Sialendoscopy and sialendoscopically-assisted operations in the treatment of lithiasis of the submandibular and parotid glands: our experience of 239 cases [J]. Br J Oral Maxillofac Surg, 2016, 54(7): 767-771.

[2] Panuganti BA, Baldassarre RL, Bykowski J, et al. Chronic sialadenitis with sialolithiasis associated with parapharyngeal fistula and tonsillolith[J]. Radiol Case Rep, 2017, 12(3): 519-522.

[3] Koch M, Schapher M, Mantsopoulos K, et al. Intraductal pneumatic lithotripsy after extended transoral duct surgery in submandibular sialolithiasis[J]. Otolaryngol Head Neck Surg, 2019, 160(1): 63-69.

[4] Stelmach R, Pawłowski M, Klimek L, et al. Biochemical structure, symptoms, location and treatment of sialoliths[J]. J Dent Sci, 2016, 11(3): 299-303.

[5] Arslan S, Vuralkan E, Cobanog B, et al. Giant sialolith of submandibular gland: report of a case[J]. J Surg Case Rep, 2015, 2015(4): 1-3.

[6] Krishnappa BD. Multiple submandibular duct (wharton′s duct) calculi of unusual size and shape[J]. Indian J Otolaryngol Head Neck Surg, 2010, 62(1): 88-89.

[7] Sigismund PE, Zenk J, Koch M, et al. Nearly 3000 salivary stones: some clinical and epidemiologic aspects[J]. Laryngoscope, 2015, 125(8): 1879-82.

[8] 刘继明, 彭铁男, 孙宏晨, 等. 下颌下静脉畸形伴多发静脉石误诊为涎石病1例[J]. 实用口腔医学杂志, 2014, 30(5): 718-720.
Liu JM, Peng TN, Sun HC, et al. Submandibular venous malformation with multiple phleboliths misdiagnosed as sialolithiasis: report of a case[J]. J Pract Stomatol, 2014, 30(5): 718-720.

[9] Eivazi B, Fasunla AJ, Güldner C, et al. Phleboliths from venous malformations of the head and neck[J]. Phlebology, 2013, 28(2): 86.

[10] Gooi Z, Mydlarz WK, Tunkel DE, et al. Submandibular venous malformation phleboliths mimicking sialolithiasis in children[J]. Laryngoscope, 2014, 124(12): 2826-2828.

[11] Prosdocimo ML, Barreto Nogueira AP, Aparecida de Albuquerque Cavalcante M, et al. Congenital dilatation of the submandibular duct[J]. Int J Pediatr Otorhinolaryngol, 2018, 113: 16-18.

[12] 张志愿. 口腔颌面外科学[M]. 7版. 北京:人民卫生出版社, 2012: 347-359.
Zhang ZY. Oral and maxillofacial surgery[M]. 7 th ed. Beijing: People's Medical Publishing House, 2012: 347-359.

[13] 薛传鹏, 薛青, 王晓武. 颌下腺导管囊肿误诊浅析[J]. 全科口腔医学杂志, 2018, 5(32): 8-10.
Xue CP, Xue Q, Wang XW. Analysis of the misdiagnosis on Wharton's Duct Cyst[J]. Gen J Stomatol, 2018, 5(32): 8-10.

[14] Juul ML, Wagner N. Objective and subjective outcome in 42 patients after treatment of sialolithiasis by transoral incision of Warthon's duct: a retrospective middle-term follow-up study[J]. Eur Arch Otorhinolaryngol, 2014, 271(11): 3059-3066.

[15] Chandra SR. Sialoendoscopy: review and nuances of technique[J]. J Maxillofac Oral Surg, 2019, 18(1): 1-10.

（编辑　罗燕鸿，刘曙光）